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Long-term visual and treatment outcomes of whole-population pre-school visual screening (PSVS) in children

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1 SUMMARY

2 What was known before

- 3 • Pre-school visual screening is effective in identifying children at risk of amblyopia and
4 is recommended to be offered for all children aged 4 to 5 years.
- 5 • Children who are socioeconomically deprived and those who come from homes that
6 require high levels of social care input are more likely to fail visual screening.

7 What this study adds

- 8 • Long term outcomes of orthoptic delivered preschool visual screening demonstrate
9 no difference in best corrected visual acuity (BCVA) and/or binocular vision (BV)
10 outcomes based on socioeconomic deprivation alone – compliance with hospital
11 attendance rates is more critical.
- 12 • Children from homes where extra social care support is required attend less well and
13 are more likely to have poorer long-term visual outcomes.

14

15 Title: Long-term visual and treatment outcomes of whole population Pre-school
16 Visual Screening (PSVS) in children: a longitudinal, retrospective, population-based
17 cohort study

18 Running title: Long-term outcomes of Pre-School Visual Screening

19

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44

45 **ABSTRACT**

46 **Background**

47 This study reports the long-term visual and treatment outcomes in a whole-population,
48 orthoptic-delivered Pre-school Visual Screening (PSVS) programme in Scotland and further
49 examines their associations with socioeconomic backgrounds and home circumstances.

50

51 **Methods**

52 Retrospective case review was conducted on 430 children who failed PSVS. Outcome
53 measures included best corrected visual acuity (BCVA), severity of amblyopia (mild,
54 moderate and severe), binocular vision (BV) (normal, poor and none), ophthalmic diagnosis
55 and treatment modalities. Parameters at discharge were compared to those at baseline and
56 were measured against the Scottish Index of Multiple Deprivation (SIMD) and Health Plan
57 Indicator (HPI), which are indices of deprivation and status of home circumstances.

58

59 **Results**

60 The proportion of children with amblyopia reduced from 92.3% (373/404) at baseline to
61 29.1% (106/364) at discharge ($p<0.001$). 80.0%(291/364) had good BV at discharge
62 compared to 29.2%(118/404) at baseline ($p<0.001$). Children from more socioeconomically
63 deprived areas (OR 2.19, 95% CI 1.01-4.30, $p=0.003$) or adverse family backgrounds (OR
64 3.94, 95% CI 1.99-7.74, $p=0.002$) were more likely to attend poorly and/or become lost to
65 follow-up. Children from worse home circumstances were 5 times more likely to have
66 residual amblyopia (OR 5.37, 95%CI 3.29-10.07, $p<0.001$) and 3 times more likely to have
67 poor/no BV (OR 3.41, 95%CI 2.49-4.66, $p<0.001$) than those from better home
68 circumstances.

69

70 **Conclusion**

71 Orthoptic-delivered PSVS is successful at screening and managing amblyopia. Children
72 from homes requiring social care input are less likely to attend and are more likely to have
73 poorer visual outcomes.

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75 **Keywords: Vision disorders, Refractive errors, Ocular motility disorders, Paediatrics,**
76 **health care economics**

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92 INTRODUCTION

93 Amblyopia is the commonest vision deficit in children in the United Kingdom and is
94 recognised to negatively impact the development of binocular vision (BV) and stereopsis.[1-
95 4]

96 The pre-school milestone (age 4-5 years) is considered the most effective time to perform
97 vision screening.[5,6] Binocular function develops from the age of 3 to 4 months and fully
98 matures by the age of 8 to 9 years.[7] Although amblyopia screening is recommended by the
99 National Screening Committee and the Hall (Four) Report,[8,9] its implementation has not
100 been without considerable variation in terms of delivery policies, screening uptake and
101 diagnostic pathways across the United Kingdom.[10,11] In view of the heterogeneity of
102 existing screening programmes and scarcity of evidence on treatment outcomes, there is a
103 need for population-based studies of long-term screening outcomes.[3,11,12]

104 The PSVS in Tayside is a whole population orthoptic-delivered programme for 4 to 5-year-
105 old children. Previously we reported the increased likelihood of failing screening for children
106 who are socioeconomically deprived and those who come from high risk homes where social
107 care input is required.[13] The aim of this current study is to report the long-term visual
108 outcomes of these children and to examine these with regard to socioeconomic and family
109 circumstances.

110

111 METHODS

112 Setting and study design

113 Details of the PSVS offered across Tayside, East of Scotland were reported in our previous
114 study.[13] Screening is delivered by orthoptists and when a child fails screening, he or she is
115 referred for repeat orthoptic assessment, cycloplegic refraction and fundus examination. The
116 vision standard to pass PSVS is best corrected visual acuity (BCVA) of ≤ 0.2 logMAR on

crowded Keeler test with each eye, or ≤ 0.1 logMAR with crowded Kay pictures if letter testing is not achieved. Children with significant refractive error are prescribed glasses and reviewed in the orthoptic clinic after up to 16 weeks; amblyopia therapy, if required, includes occlusion or atropine penalization. Children who are treated for amblyopia are examined every 6-8 weeks until BCVA improves to an age-appropriate level or is stable and deemed unlikely to improve further.

The study group comprised the same 523 children who failed PSVS from a total number of 4365 (11.9%) children screened between March 2010 and February 2011 (as in our previous study).[13] A retrospective case review was performed to identify visual outcomes for each child up until either their final discharge visit, or most recent outpatient visit whichever came later. Outcome measures included BCVA, refractive status, residual amblyopia (if any) and BV. As we have previously reported on the rate of screening uptake and reasons for failing screening, these are not included in our current report.[13] In the event when a child had bilateral amblyopia, data from the worse seeing eye was used to avoid inter-eye correlations. Given the study was not conducted in a trial setting, there is no standard operating procedures for orthoptic appointments as the orthoptists work as autonomous practitioners who pick the most appropriate test for examination depending on the child's level of cooperation and vision on the day of visit.

Ninety-three children either did not attend any clinic appointments after the screening event or no follow-up data were available, leaving 430 children with clinical information on both their screening and subsequent follow-up appointments. Children who failed to attend were offered two further appointments before being discharged via letter to their general practitioner (GP) and health visitor (HV). This is summarised in **figure 1**. Of the 430 children who were seen after the screening event, 40 failed to attend before treatment was completed. This group of children was categorised as poor attenders and their last recorded visual outcomes were used for a separate analysis.

144 **Definitions**

145 *Scottish Index of Multiple Deprivation (SIMD)*

146 The SIMD 2012 (Scottish Government) is a multidimensional indicator, taking account of
147 seven domain scores to produce an overall deprivation score for different postcodes. In our
148 series of case studies, we have divided the SIMD into two distinct groups to examine the link
149 between extreme deprivation and long term visual outcomes: Quintile 1 (0-20% most
150 deprived) and Quintiles 2-5 (20-100% least deprived).

151

152 *Health Plan Indicator (HPI)*

153 This is a unique code given by the assigned HV of every child in the UK based on a
154 comprehensive assessment of the needs of children and individual family circumstances.
155 Three HPI codes were used at the time of this study and they, in order of increasing need for
156 input from health and social services are Core (C), Additional (A) and Intensive (I). A child
157 from a stable home with no concerns would be assigned 'Core' and receive HV and GP
158 input; a child from an unstable home, for example with substance abuse problems, could be
159 assigned 'Intensive' and subsequently receive more input from health and social services.
160 The HPI is the only formally applied measure of the stability and security of a child's home
161 environment, it is widely used and well validated.

162

163 *Strabismus*

164 Full orthoptic assessment of strabismus was undertaken, strabismus included any constant
165 or intermittent heterotropia, and micro-strabismus.

166

167

168 *Amblyopia*

169 We defined amblyopia as BCVA ≥ 0.2 logMAR in the amblyopic eye and/or interocular
170 difference of 3 or more logMAR lines. We excluded children with co-existing ocular
171 abnormalities precluding normal vision. For children with bilateral amblyopia, visual acuity of
172 the worse eye at baseline was used for comparison purposes.

173 We categorised amblyopia severity into three categories based upon the worse eye BCVA
174 using the US Pediatric Eye Disease Investigator Group (PEDIG) definitions[14]; Mild: better
175 than 0.3 logMAR; Moderate: 0.3-0.7 logMAR; Severe: worse than 0.7 logMAR.

176

177 *Binocular vision (BV)*

178 At the screening event, the orthoptists indicated “yes” or “no” for BV based on a child’s
179 response to a 15 Δ prism reflex test and screening TNO plates. BV was further assessed at
180 all clinic appointments. Frisby stereo-acuity test was used to assess stereopsis and Wirt fly
181 was used if Frisby was not achieved. Motor fusion was assessed using the 15 or 20 Δ base
182 out test. When BV was not performed at the discharge visit, the final recorded BV closest to
183 a child’s discharge visit was used for comparison with the BV recorded at the first orthoptic
184 visit which was subsequent to refraction and fundus check.

185 The range of BV was divided into three groups. Normal BV: Stereopsis better than 170
186 seconds of arc and the ability to overcome a prism; Poor BV: stereopsis of 170 -600 seconds
187 of arc irrespective of ability to overcome a prism or the inability to overcome a prism
188 irrespective of level of stereopsis; No BV: Stereopsis poorer than 600 seconds of arc and the
189 inability to overcome a prism.

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Statistical analysis

SPSS statistical package (IBM SPSS Statistics for Windows, V.19.0, IBM Corp, Armonk, New York, USA) was used for data analyses. Chi-squared test (X²) was used to calculate the association between categorical variables and socioeconomic background as well as home circumstance based on SIMD and HPI respectively. One-way analysis of variance was used to assess the difference in continuous variables among different subgroups. Hypothesis test of the equality of two proportions were used to compare proportions of amblyopia and BV. Mixed regression model was used to evaluate the relationship between BCVA and BV at discharge. All analyses were done with 95% confidence interval, and a p-value of ≤ 0.05 considered statistically significant.

RESULTS

Study group and background demographics

Results of the first clinic appointment (repeat orthoptic assessment, refraction and examination) were available for 430 of the 523 children (82.2%) who failed screening. The remaining 93 of the 523 children (17.8%) either did not attend their referral appointment from screening (Baseline visit) or there were no data available.

Of those who did attend their first appointment the attendance rate for follow-up at the eye clinic was 90.7% (390/430). Figure 1.

Background demographic and pattern of attendance to follow-up clinic visits are summarised in **Table 1**.

217 **Poor attenders**

218 Forty of the 430 children (9.3%) with follow-up results were categorised as poor attenders.

219 Mean (\pm standard deviation) age at discharge for this group was 6.2 ± 1.2 years old; their

220 mean duration of follow-up was 26.5 ± 10.5 months.

221 Sixteen (40.0%) of the 40 poor attenders were from the 0-20% most deprived socioeconomic

222 group. The odds of children from the 0-20% most deprived socio-economic group of having

223 poor attendance were twice as high as for those from the 20-100% least deprived

224 socioeconomic group (OR 2.19, 95% CI 1.01-4.30, $p=0.003$).

225 Eighteen (45.0%) of the 40 poor attenders were from a family assigned as either “Intensive”

226 (I) or “Additional” (A). The odds of children from HPI groups I and A of attending poorly were

227 four times higher than children from HPI group C (OR 3.94, 95% CI 1.99-7.74, $p=0.002$).

228

229 **Ophthalmic diagnosis**

230 Of the remaining 390 children who were regular attenders, 387 (99.2%) were discharged

231 from the clinic after a mean follow up time of 19.7 ± 5.8 months.

232 Twenty six of the 430 children (6.0%) who met the referral criteria were discharged after one

233 to two visits if their vision proved to be normal, these children were classed as false positives

234 and excluded from the outcome data. A further 31 (7.2%) children were reviewed at least

235 three times without any active intervention because they had reduced vision but no evidence

236 of refractive error or pathology and eventually they demonstrated a satisfactory level of

237 vision (VA $< \log\text{MAR } 0.2$). These children were grouped as “visually immature” because with

238 age and repeated practice at the assessment they were able to achieve normal vision.

239 These children underwent cycloplegic refraction and dilated fundoscopy by a paediatric

240 ophthalmologist or hospital optometrist, as all our children do, and no pathology was found.

241 **Management**

Two hundred and fifty-four children were prescribed glasses; this was the sole intervention for 173 of the 390 children (44.4%) who attended regularly. 102 (26.1%) were treated with occlusion. Six children (1.5%) received atropine penalisation, 4 of whom had adjuvant patching. Two refused patching.

A total of twenty-four (6%) children were recorded as being non-compliant with either glasses (n=4) or occlusion (n=20), of which 10 were poor attenders and were lost to follow-up. Sixteen (66.7%) of these children were from a family assigned as “Intensive” or “Additional”. (OR 9.97, 95% CI 0.23-0.71, $p<0.001$). Five (20.8%) were from the 0-10% most socioeconomically deprived background.

Ten children (2.1%) received surgical correction for strabismus, for whom the mean overall length of follow-up in total was 3.08 ± 1.40 years.

Amblyopia

The proportion of children with amblyopia at baseline and the final visit for both poor and regular attenders is shown in **figure 2**.

At baseline visit, 373 children (92.3%) had amblyopia. 62/373 (16.6%) were categorised as mild, 273/373 (73.1%) moderate and 38/373 (10.2%) severe.

For poor attenders (N=40) who were lost to follow-up, 72.5% had their last measured BCVA recorded as meeting the amblyopia threshold; of these 6 (15.0%) were categorised as mild, 20 (50.0%) moderate and 3 (7.5%) severe.

For the remaining 364 children who attended clinic regularly, 70.9% children (n=258) had BCVA better than 0.2 logMAR at discharge. Difference between the proportion of children with amblyopia at baseline and at discharge was statistically significant ($p<0.001$).

The odds of having amblyopia at the baseline clinic visit was 29 times higher than at the point of discharge (OR 29.29, 95% CI 7.84-26.14, $p<0.001$). The odds of having residual

amblyopia for poor attenders was significantly higher than children who attended follow-up appointments regularly (OR 6.42, 95% CI 4.25-10.56, $p<0.001$).

Binocular vision

At the point of screening 161 of 430 children (37.4%) who were referred had their BV recorded as “no”. At baseline orthoptic clinic visit, after refraction and fundus examination, 118/404 (29.2%) had good BV, 185/404 (45.8%) had poor BV and 101/404 (25.0%) were recorded as no BV. Of the regular attenders, at discharge, 291/364 (79.9%) had good BV, 49/364 (13.5%) had poor BV and 24/364 (6.6%) had no BV. The distribution of BV pattern proportion at baseline and at the final visit is summarised in **figure 3**.

The difference between the proportion of children with good BV at baseline and at discharge was statistically significant ($p<0.001$). The odds of having good BV at discharge for the regular attenders was 7 times higher than that at baseline (OR 9.7, 95% CI 0.62-1.10, $p<0.001$). There was a positive association between BCVA and BV at final discharge ($r=0.88$, 95% CI 0.76-0.91, $p<0.001$).

Of the 40 poor attenders, at baseline clinic visit, 8 (21.1%) had good BV, 21 (55.3%) had poor BV and had 9 (23.7%) had no BV. Twelve (31.6%) were last recorded as having good BV, 18 (47.4%) had poor BV and 8 (21.1%) had no BV.

The difference between the proportion of children having poor/no BV among the poor attenders compared to the regular attenders is significant ($p<0.001$).

Comparison of final visual outcome based on SIMD and HPI

The relationship between socioeconomic background (SIMD), home circumstance as indicated by HPI and adverse visual outcome for children who attended well ($n=364$) was examined (**Table 2**). Results were independent of gender and ethnicity for these children.

There was no statistical difference in the odds of children from the 0-20% most deprived socioeconomic background having poorer visual outcomes (final BCVA worse than logMAR 0.2, improvement of BCVA less than logMAR 0.2 and poor or no BV) compared to children from the 20-100% least deprived socioeconomic background. ($p=0.745$, $p=0.710$, $p=0.219$ respectively).

However, children from HPI groups I and A were 5 times more likely to have a final BCVA worse than 0.2 logMAR (OR 5.37, 95%CI 3.29-10.07, $p<0.001$) and 3 times more likely to have poor or no BV (OR 3.41, 95%CI 2.49-4.66, $p<0.001$) compared to children from a family assigned as "Core".

DISCUSSION

Overall the children in our real life cohort responded well to amblyopia treatment, with 70.9% of good attenders achieving a BCVA of better than 0.2 logMAR and 61.7% achieving an improvement of at least 0.2 logMAR. The proportion of children with moderate to severe amblyopia reduced from 77.0% at baseline to 8.7% at discharge. The magnitude of this improvement was comparable to that observed in randomised controlled trials such as the ALSPAC and PEDIG studies.[15,16]

Our results also demonstrated an increase in the proportion of children with good BV from 29.2% at baseline to 79.9% at discharge. Previous studies have shown that BV can improve following treatment of amblyopia.[17-19] Our study supports these findings, including in those who had intermittent heterotropias and micro-strabismus.

This study found that children from more deprived socioeconomic backgrounds and those from families requiring more social care input (HPI) are more likely to have poor attendance.

Analysis of the visual outcomes for poor attenders in our study showed that they were 6 times more likely to have residual amblyopia and almost 10 times more likely to have poor or no BV compared to regular attenders. Children who were poor attenders and those who became lost to follow-up record a relatively earlier last visit during their treatment, which meant they had fewer attempts to have improved visual acuity and less time to be treated in a closely monitored specialist setting. It is possible that poorer health seeking behaviour among parents who require social care input adversely impacts on the attendance rate of their children as they are less likely to engage with health services.[20] The attendance rate for follow-up eye clinic appointments in our study sits around 90.7%, which is higher than most other studies.[15,21]

Our results have demonstrated that irrespective of a child's socioeconomic background, with regular follow-up, intensive treatment and good compliance, children from more deprived backgrounds have similarly good visual outcomes compared to less deprived children. This is an important finding as our initial study found that children who were from deprived backgrounds were more likely to fail screening.[13] In this study, children from less stable home circumstances who required "Intensive" and "additional" support were 4.5 times more likely to have a worse final BCVA and 3 times more likely to have poor or no BV compared to children from the "core" group. This study also reported a similar association between worse home circumstances and screening outcomes.[13] Children from the "Intensive" and "additional" group were 10 times more likely to be treatment non-compliant, irrespective of socioeconomic background.

The reasons for this difference in screening failure rates are not known but it has been theorised that poorer prenatal/antenatal care [22, 23] associated with increased rates of maternal smoking, alcohol and drug intake [24, 25, 26, 27] which are commoner in deprived areas [28] may be significant contributors. This current study suggests that, if these factors are indeed relevant, they are reversible with adequate treatment. Comprehensive screening

to pick up these most vulnerable children is essential and it must be followed up by methods to encourage treatment compliance.

One limitation of our study was the retrospective nature of the data collection, but the benefit of this methodology is that the observational findings are representative of the real-life situation. The percentage of children lost to follow-up (9.3%) was slightly higher than other studies.[11,15-16] However, our results have shown that the majority of the poor attenders were from more socioeconomically deprived and adverse family backgrounds and that the home circumstances associated with poor attendance have the most impact on the outcome. Hence although this is a form of bias, it contributes to a possible underestimation of the negative impact of deprivation on the final visual outcome.

This study reports the treatment and visual outcomes of a whole population orthoptic-delivered preschool visual screening service. It identified that attendance is the key to the final visual outcome for children; children from deprived/high risk homes were much more likely to not attend appointments and did not do well. It is crucial for children who are already being brought up in a challenging environment that the screening system supports them and their families, in order that they may have the same successful outcomes as their more fortunate peers.

366 **CONTRIBUTORS**

367 Data were collected by YN, UOC and CG. Statistical analysis was performed by YN. YN
368 produced the initial draft manuscript, all authors contributed to the revision and UOC and
369 CJM prepared the final draft.

370 **COMPETING INTERESTS**

371 None declared.

372

373 **FUNDING**

374 None

375

376 **ETHICS APPROVAL**

377 Ninewells Hospital Orthoptic and Ophthalmology departments have ongoing Caldicott
378 guardianship approval for analysis and review of preschool vision screening outcomes.

379

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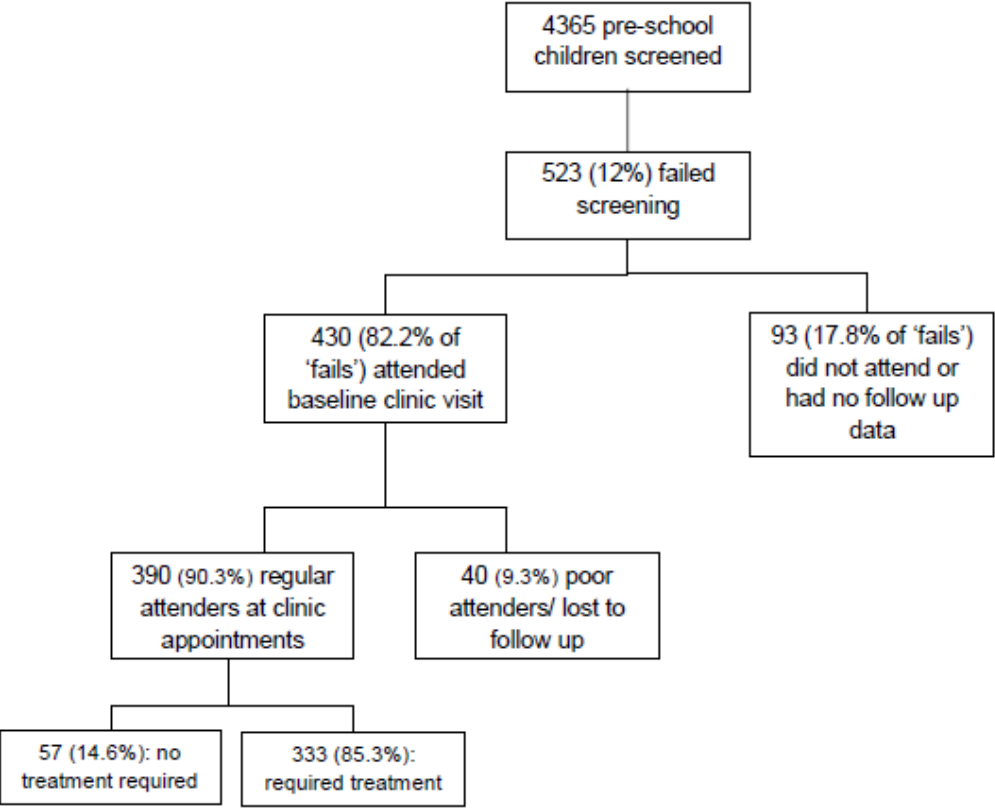
457 **Titles and legends to figures**

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459 Figure 1: Flow chart summarising the number of children who underwent Pre-school Visual
460 Screening (PSVS) and number of children included in the final analysis of this study.

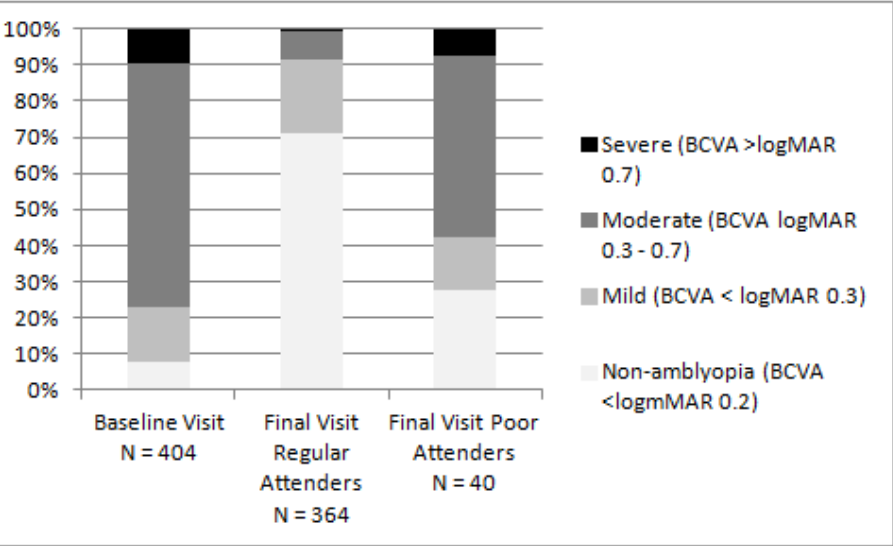
461 Figure 2: This graph shows the distribution of amblyopia based on the level of severity (mild,
462 moderate and severe) at baseline and final visit for regular and poor attenders.

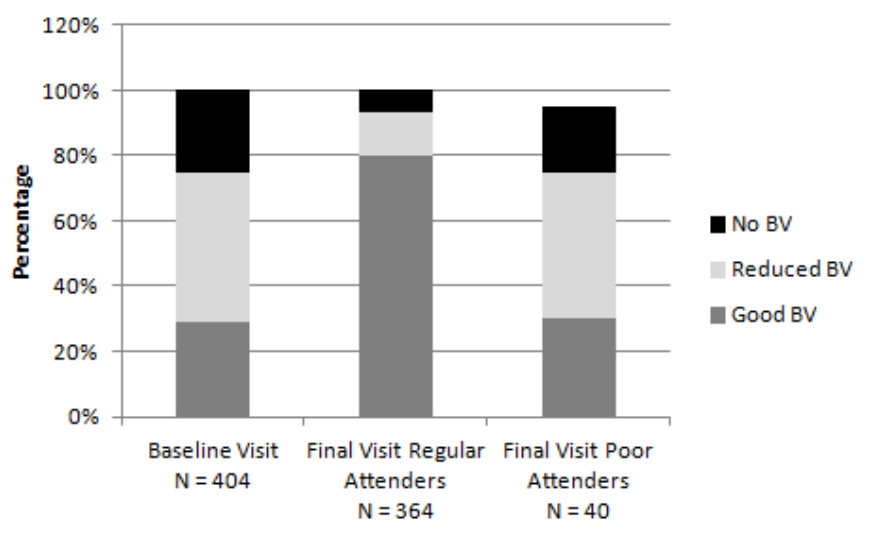
463 Figure 3: This graph shows the distribution of binocular vision (BV) at baseline and final visit
464 for regular and poor attenders.



Scottish Index of Multiple Deprivation (SIMD)		No. (%) of children (n=430)
	Quintile 1 (Most deprived)	107 (24.9%)
	Quintile 2	62 (14.4%)
	Quintile 3	82 (19.1%)
	Quintile 4	130 (30.2%)
	Quintile 5 (Least deprived)	49 (11.4%)
Health Plan Indicator (HPI)		
	Intensive (I)	22 (5.1%)
	Additional (A)	63 (14.7%)
	Core (C)	345 (80.2%)
Attendance		
	Regular attender	390 (90.7%)
	Poor attender	40 (9.3%)
Gender		
	Male	207 (48.1%)
	Female	223 (51.9%)
Ethnicity		
	Caucasian	421 (97.9%)
	Others	9 (2.1%)

Table 1: This table details the background socioeconomic status, health plan indicator and pattern of attendance to follow-up clinic for study population





	Scottish Index of Multiple Deprivation (SIMD)				Health Plan Indicator (HPI)			
	Quintile 1 (0-20% most deprived)		Quintile 2-5 (20-100% Least deprived)		Intensive (I) and Advanced (A)		Core (C)	
	n (%)	OR (95% CI)	n (%)	p-value	n (%)	OR (95% CI)	n (%)	p-value
Final BCVA >logMar 0.2	22 (24.2%)	0.82 (0.48-1.40)	84 (28.1%)	0.745	40 (59.7%)	5.37 (3.29-10.07)	66 (20.4%)	<0.001
Improvement of BCVA <logMar 0.2	31 (34.0%)	1.11 (0.68-1.82)	95 (31.8%)	0.710	25 (37.3%)	1.31 (0.76-2.27)	101 (31.3%)	0.264
Poor / No BV	21 (23.0%)	1.40 (1.19-3.94)	52 (17.4%)	0.219	25 (37.3%)	3.41 (2.49-4.66)	48 (14.9%)	<0.001

Table 2: A comparison of the odds of children having poorer visual outcomes (final BCVA more than 0.2 logMAR, improvement of BCVA less than 0.2 logMAR and reduced/no binocular vision) based on recorded SIMD and HPI at discharge.